

CASE REPORT

Mature Solid Teratoma of the Fallopian Tube Associated with Uterine Leiomyomas

Tai-Jong Chao¹, Joe Chao², Long-Jin Kuan³, Yiu-Ta Li³, Tseug-Cheng Kuo³,
Yun-Ching Chang⁴, Shin Nieh^{4*}

¹Department of Pathology, ²Research Center, and ³Department of Obstetrics and Gynecology, Kuo General Hospital, and ⁴Department of Pathology and Graduate Institute of Pathology, National Defense Medical Center and Tri-Service General Hospital, Taipei, Taiwan, R.O.C.

Benign solid teratomas within the fallopian tube are rare. The incidence of this kind of benign solid teratoma of the fallopian tube associated with intrauterine leiomyomas is extremely low and may complicate the clinical manifestations. Here, we report a 40-year-old female with periodic lower abdominal pain and hypermenorrhea who was found to have 3 nodules within the uterine myometrium and left distended fallopian tube. Final pathology revealed benign solid teratoma of the fallopian tube associated with 3 intrauterine leiomyomas. Primary teratoma of the fallopian tube is extremely uncommon. Gynecologic oncologists should be aware of the possibility of this disease entity when making differential diagnoses. [*J Chin Med Assoc* 2008;71(8):425–427]

Key Words: benign solid teratoma, differential diagnoses, fallopian tube, leiomyoma

Introduction

The finding of a benign solid teratoma within the fallopian tube is rare. The incidence of solid mature teratoma of the fallopian tube associated with intrauterine leiomyoma is extremely low. Most cystic teratomas are intraluminal and located in the isthmus and ampullary regions of the fallopian tube. Patients tend to be nulliparous or have parity of less than 2. None of the reported cases were diagnosed preoperatively. Tumor rupture with signs of peritonitis has been reported.¹ They were all incidental findings during an operation or examination of a surgical specimen.

Mature solid and cystic teratomas (dermoid cysts) are known to occur most commonly in the gonads. Extragonadal occurrences are observed primarily in the sacrococcygeal region, mediastinum, retroperitoneal cavity, cranial cavity or in the neck area.^{2,3} Cystic teratomas are usually found in young women during their active reproductive years. Discovering a 4 × 2 × 2 cm mostly solid tumor arising from the muscular layer of our patient's left fallopian tube associated with intrauterine leiomyomas was a rare occurrence and prompted us to make this report. The pathogenesis of

teratoma of the fallopian tube remains unclear. We consider the origin to be from germ cells, as teratoma is found most frequently in the gonads and along the known pathway of germ cell migration or from the meiotic germ cells. The karyotype of all benign teratomas is 46,XX.^{2,4–7}

Case Report

A 40-year-old patient, gravida 6, para 2, abortion 4, complained of severe dysmenorrhea with periodical lower abdominal pain, hypermenorrhea, and occasional blood clots of a few months' duration. She denied fever or the passage of tissue from the uterus. Symptoms did not improve after taking Azulensol 2 mg, Rowaprxin 2 mg and Strocaine 10 mg. She later visited our outpatient department and was admitted for further evaluation. Her past medical history was positive for an appendectomy 3 years earlier. She denied previous pelvic inflammatory disease. Her family history was negative for any hereditary disease.

Physical examination revealed an oriented woman in no distress with a temperature of 37.2°C, blood



*Correspondence to: Dr Shin Nieh, Department of Pathology and Graduate Institute of Pathology, Tri-Service General Hospital, 325, Cheng-Kung Road, Section 2, Neihu, Taipei 114, Taiwan, R.O.C.
E-mail: niehshin1014@yahoo.com.tw • Received: September 28, 2007 • Accepted: April 1, 2008



Figure 1. Sonographic examination of the uterus shows 3 nodules within the uterine myometrium.

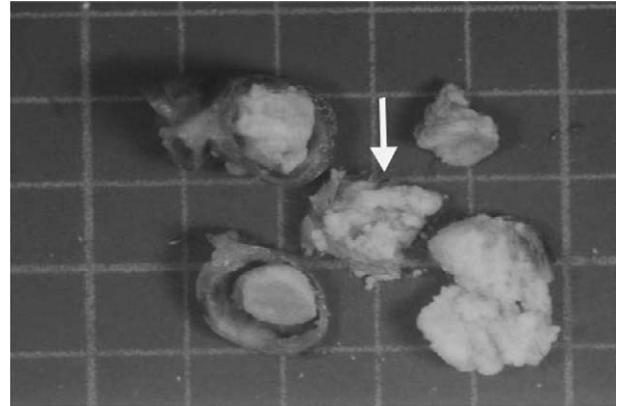


Figure 3. Gross findings of the tumor show both solid part and cystic content. There is greasy, sebaceous material and sparse black hairs (arrow).



Figure 2. Operative finding shows that the tumor had induced a sausage-like swelling in the left fallopian tube.

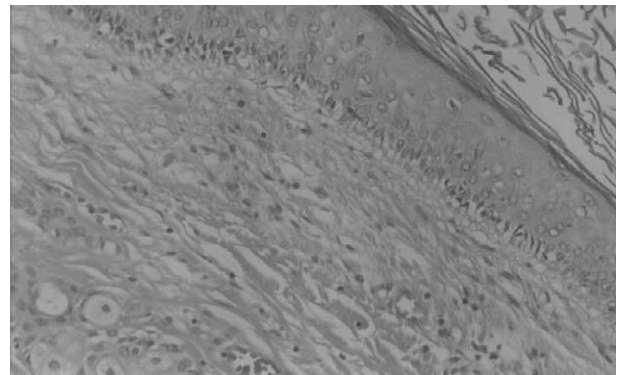


Figure 4. Microscopic finding of mature teratoma demonstrates a cyst lined entirely by well-differentiated keratin-producing squamous epithelium (hematoxylin & eosin, 400 \times).

pressure of 118/75 mmHg, pulse of 78 beats/minute, and respiration of 18 breaths/minute. She had no tenderness or palpable masses on abdominal examination. Pelvic examination revealed 2 mL of old blood in the vaginal vault. She had cervical erosion. Sonographic examination showed 3 nodules within the uterine myometrium measuring about 2.8×2.2 cm, 2.4×2.2 cm and 1.6×1.2 cm (Figure 1). Laboratory examination results were within normal limits. Quantitative β -human chorionic gonadotropin was normal (<5 IU/mL). The patient underwent surgery with a preoperative diagnosis of intrauterine leiomyomas and adenomyosis was also put into consideration for differential diagnosis. At the time of the operation, the uterus was found to be enlarged due to 2 subserosal leiomyomas and an intramural leiomyoma. The left fallopian tube appeared to be predominantly solid and distended, containing cream-colored sebaceous material and hair (Figure 2). The contralateral adnexa and left ovary were normal. The patient

subsequently underwent total abdominal hysterectomy with left salpingectomy.

Gross examination of the left fallopian tube revealed a $4.0 \times 2.2 \times 1.2$ cm solid tumor. It contained a yellowish, cheesy, sebaceous material and black hair (Figure 3). Cross section revealed a thin wall lined by an opaque yellowish, gray-white wrinkled apparent epidermis. Within the wall was calcification. The distal and proximal portions of the tubal lumen appeared normal. On microscopic examination, the left fallopian tube showed mature solid teratoma with focal cystic change. The solid portion of the fallopian tube contained a plug of fatty, cheesy, waxy tissue. The wall of the tube was composed of well-differentiated squamous epithelium, hair follicles or shafts, underlying stratified squamous cells, sebaceous glands, and other skin adnexal structures (Figure 4).

Close follow-up sonographic surveillance and laboratory examination (β -human chorionic gonadotropin <5 IU/mL; α -fetoprotein <20 ng/mL) proved normal.

The patient had no symptoms or signs at the 4-month follow-up.

Discussion

The benign teratoma of the fallopian tube is composed of recognizable tissues of ectodermal, mesodermal and endodermal origin, in any combination.² The term dermoid cyst was coined over 160 years ago. It refers to a mature teratoma that is composed predominantly of a cyst lined entirely or partly by well-differentiated keratin-producing squamous epithelium, which emerges from the tubal wall, not continuous with the tubal epithelium. It initially presents as of mesodermal origin with abundant mesenchymal stroma, but eventually develops both endodermal and ectodermal derivatives with airway lining enterocytes, thyroid brain and skin appendages. Primary teratoma of the fallopian tube is extremely uncommon. At the present time, only about 58 cases have been reported in the literature, including 1 from Taiwan.^{8,9} The majority of the cases occurred in patients in their 40s, and were cystic, showing great variation in size. The first case in Taiwan, which we have described, was a solid teratoma in the intraluminal location of the fallopian tube associated with intrauterine leiomyomas. Histologically, they are similar to teratoma of the ovary and elsewhere. This 45-year-old woman underwent laparoscopy for intra-uterine leiomyoma clinically diagnosed after a significant period of abdominal pain and dysmenorrhea, with ultrasound findings of leiomyoma of the uterus. The benign teratoma of the fallopian tube was distended by cheese-like material, found during surgery. Left salpingectomy was performed. The finding was confirmed by pathology. Herein, we have presented the first case of benign solid teratoma of the left fallopian tube associated with intrauterine leiomyomas.

In conclusion, benign teratomas are the most common of all ovarian neoplasms, and represent a diverse

group of tumors that may develop at other sites. They develop from a totipotential stem cell. Most importantly, the diversity of teratoma behavior probably reflects the different biological potentials of various stem cells, including germ cells and pluripotent embryonic cells. Benign teratoma of the fallopian tube associated with intramural leiomyoma is extremely rare. If the tumor is not large enough, preoperative diagnosis is difficult. Prognosis is favorable following complete surgical excision. About 5–10% of dermoids (mature teratomas) undergo malignant transformation of any one of the component elements (e.g. adenocarcinoma, choriocarcinoma, thyroid carcinoma, melanoma, but most commonly squamous cell carcinoma).^{3,10}

References

1. Horn T, Jao W, Keh PC. Benign cystic teratoma of the fallopian tube. *Arch Pathol Lab Med* 1983;107:48.
2. Mutter GL. Teratoma genetics and stem cells: a review. *Obstet Gynecol Surv* 1987;42:661–70.
3. Mazzarella P, Oksggki T, Richart RM. Teratoma of the uterine tube: a case report and review of the literature. *Obstet Gynecol* 1972;39:381–8.
4. Norris HJ, Zir HJ, Barson WL. Immature (malignant) teratoma of the ovary: a clinical and pathological study of 58 cases. *Cancer* 1976;37:2359–72.
5. Corfman PA, Richart RM. Chromosome number and morphology of benign cystic teratoma. *N Engl J Med* 1964;271:1241–4.
6. Rashad MN, Fathalla RM, Kerr MG. Sex chromatin and chromosome analysis in ovarian teratomas. *Am J Obstet Gynecol* 1966;96:461–5.
7. Linder D, McCaw BK, Hecht F. Pathogenic origin of benign ovarian teratoma. *N Engl J Med* 1975;292:63–6.
8. Johson C, Hansen KA. Mature cystic teratoma of the fallopian tube. *Fertil Steril* 2006;86:995–6.
9. Hseih CS, Cheng GF, Liu YG, Han CP, Chen SS. Benign cystic teratoma of unilateral fallopian tube associated with intra-uterine pregnancy. *J Chin Med Assoc* 1998;61:239–42.
10. Doss BJ, Jacques SM, Qureshi F, Chang CH, Christensen CW, Morris RT, Lawrence WD. Immature teratomas of the genital tract in older women. *Gynecol Oncol* 1999;73:433–8.